Subcutaneous Emphysema Following Nasal Positive Airway Pressure Therapy in a Patient With a History of Tongue Base Tumor Resection

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Positive airway pressure (PAP) therapy is chosen as the primary modality for most patients with obstructive sleep apnea due to its efficacy and safety. However, PAP therapy can cause potential side effects, including barotrauma, which in most cases is benign and manageable. Nevertheless, rare but serious complications, such as subcutaneous emphysema, may also occur. Here, I present a case of a middle-aged man with a history of tongue base tumor resection who developed subcutaneous emphysema in the neck following nasal PAP therapy. This case highlights the potential risk associated with previous surgical interventions and PAP therapy.

INTRODUCTION

Positive airway pressure (PAP) therapy is an effective treatment for patients with moderate to severe obstructive sleep apnea (OSA) [1]. PAP is safe, with minimal or reversible adverse effects. However, as the use of PAP devices continues to increase, it is crucial to recognize the potential side effects. Barotrauma associated with PAP therapy is known to cause dryness of the nasal and oral mucosa, sinusitis, congestion, epistaxis, and aerophagia [2]. Nevertheless, PAP therapy may lead to rare yet potentially severe complications, such as subcutaneous emphysema [3,4]. Here, I present a case of a middle-aged man who underwent tongue base tumor resection and experienced subcutaneous emphysema in the neck following nasal PAP therapy.

CASE REPORT

A 45-year-old male patient presented to a sleep clinic with complaints of snoring, witnessed apnea, dry mouth, and sleep-related choking. His body mass index was 24.2 kg/m². A Mallampati score of 3 and an Epworth Sleepiness Scale score of 13 supported a high risk of OSA. In terms of medical history, he had undergone radical dissection with selective lymph node removal and sublingual gland reconstruction due to a tongue base tumor 9 months prior but he was otherwise healthy. Diagnostic in-lab polysomnography revealed severe OSA with an apnea-hypopnea index of 46.8 events per hour and a nadir of oxygen desaturation of 64%. Nasal automatic PAP in the range of 6–12 cm H₂O was prescribed before continuous PAP (CPAP) titration. At the CPAP titration on day 25 of automatic PAP use, a pressure of...
9 cm H2O was found to be effective. However, on day 27 after automatic PAP use, the patient developed mild soreness on the tongue base, chewing discomfort, sore throat, and low-grade fever with a sensation of heat and swelling in the area from the chin to the neck. The patient immediately stopped automatic PAP, took acetaminophen for symptom relief, and visited the sleep clinic on the eighth day after discontinuation of PAP. The patient did not complain of dyspnea and reported that his neck swelling was much improved. On physical examination, his oral cavity was clear, but mild swelling on the right side of the neck was evident and multiple small nodular bubbles with crepitus were still palpable on the neck (Fig. 1), and lung sounds were normal. He denied any history of body trauma. Before the onset of symptoms, the patient was comfortable with automatic PAP, and his adherence was good. Considering the close temporal relationship between the use of PAP and the occurrence of subcutaneous emphysema and improvement of symptoms after discontinuation of PAP, a tentative diagnosis of PAP-induced subcutaneous emphysema was made, and PAP was halted until the symptoms had completely resolved. The patient was referred to the Department of Dentistry and Otolaryngology for further evaluation, which included physical examination, laryngoscopy, and computed tomography of the neck and chest, but the evaluation failed to identify the causative lesion. The subcutaneous emphysema disappeared within 2 weeks of PAP withdrawal. After 30 days, nasal CPAP therapy at 9 cm H2O was restarted, and the subcutaneous emphysema has not recurred for more than 2 years.

DISCUSSION

The presence of air or gas in the subcutaneous tissue layer refers to subcutaneous emphysema. Trauma or injury to the neck or chest can cause the airway or lungs to rupture, permitting air or gas to escape into the surrounding tissues and spread along the fascial planes of the adjacent body part [5]. Iatrogenic injury, such as mechanical ventilation or endoscopic procedures, can be another etiology [5]. Barotrauma due to high PAP is also thought to accompany [5-7]. Swelling and pain in the affected area are common symptoms, and a sore throat, wheezing, or breathing difficulty may accompany. A typical sign of physical examination with crepitus, a cracking or popping sensation felt under the skin, is diagnostic in most of the cases [5].

Although the direct cause in this case is unclear, it is likely that previous surgical sites may have acted as a conduit for the air propelled by positive pressure, contributing to the development of subcutaneous emphysema. As subcutaneous emphysema occurred with tongue and oral soreness, unidentified oropharyngeal mucosa, especially previous surgical sites, including the reconstructed sublingual gland, may be the potential sources of subcutaneous emphysema in this case. There are only two reported cases of PAP-induced subcutaneous emphysema: a 15 cm H2O CPAP user who developed subcutaneous emphysema of the face and neck after facial trauma due to falling out of bed [4], and an 11 cm H2O CPAP user who developed a periorbital subcutaneous emphysema [3]. It has been speculated that the former occurred through unidentified fracture sites and the latter occurred through the lacrimal duct.

Treatment involves addressing the underlying cause and providing supportive care. If the causative lesion is identified, surgical correction should be considered. While temporarily discontinuing PAP therapy was effective in managing this case, medical attention should be sought as it can cause respiratory issues. Potential air passages may be spontaneously closed after PAP discontinuation and a mild inflammatory reaction following subcutaneous emphysema.

PAP should be prescribed cautiously in patients with a history of oral surgical procedures and facial trauma with possible bone fractures. To my knowledge, this case presents the first report on the occurrence of subcutaneous emphysema following PAP therapy in an OSA patient with a history of surgical removal of base of the tongue tumor, highlighting the need for further
research on the potential risks associated with previous surgical interventions and PAP therapy. Resumption of PAP therapy should be approached with caution, as there is a concern about recurrence. I recommend restarting PAP therapy when the symptoms have completely resolved, gradually resuming the pressure or lowering the pressure from the previous setting, and monitoring for adverse effects or signs of recurrence. Subcutaneous emphysema may be a rare but possible adverse effect of PAP therapy, and clinicians should be aware of this potential complication.

**Ethics Statement**
This study followed the principles of the Declaration of Helsinki, and it was approved by the Inje University Busan Paik Hospital Institutional Review Board (BPIRB#: 2023-01-036). Written informed consent was obtained from the patient for publication of this case report and accompanying images.

**Availability of Data and Material**
All data generated or analyzed during the study are included in this article.

**Conflicts of Interest**
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**REFERENCES**